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Glandular Odontogenic Cyst of Maxilla: A Case Report

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ABSTRACT

Glandular Odontogenic Cyst (GOC) is a developmental odontogenic cyst with distinct clinical, radiographical and histological features. Its age predilection for occurance is in the middle aged individuals and the most common site of occurrence is the anterior mandibular region. Radiologic features of GOC are not pathognomonic. It may manifest as either unilocular or multilocular radiolucency, usually with well defined scalloped margins often crossing the midline. The locularity (unilocular or multilocular), radiodensity and border characteristics of GOC are important in the differential diagnosis. We report here the clinical, radiologic, histopathologic features and one year follow up of glandular odontogenic cyst occurring in the posterior maxilla which is relatively rare.

KEY-WORDS: Developmental odontogenic Cyst, Glandular Odontogenic Cyst, Sialo Odontogenic Cyst.

INTRODUCTION

Glandular odontogenic cyst is a rare cyst of developmental odontogenic origin [1,2]. **Padayachee** and **Van Wyk** in 1987 speculated the possibility of salivary gland origin, reported two multilocular mandibular lesions with histopathological features of botryoid odontogenic cyst and mucoepidermoid tumour[3,4]. GOC is defined as a cyst arising in the tooth-bearing areas of the jaws and characterized by an epithelial lining with cuboidal or columnar cells both at the surface and lining or cyst-like spaces within the thickness of the epithelium [5].

GOC has a frequency rate of only 0.012% to 1.3% of all jaw lesions[3]. It is therefore, seldom suspected on clinical and radiological examination.

The recurrence rate of GOC ranges between 21% and 55%[1]. The aggressive nature of GOC's in maxilla makes it distinct from other cystic lesions of the jaw bones thus diagnosing the disease prior to surgical intervention is essential in this regard.

The present article reports a case of GOC involving the maxillary posterior region.

CASE HISTORY

A 15 year old female patient reported at the Department of Oral Medicine and Radiology with complaint of a painless swelling in the left middle third of the face present since 2 – 3 months. It had gradually increased its rate of growth in the last one month. There was no history of toothache or pus discharge associated with the swelling. Medical history of patient was unremarkable. Facial asymmetry owing to a solitary, diffuse, oval swelling was noted over the left middle third region of the face (fig 1). There was obliteration of the naso labial fold with normal overlying skin. Swelling was non-tender and bony hard on palpation. Intra oral examination revealed grossly decayed 26 along with obliteration of the buccal vestibule in relation to the teeth 23 to 27 (fig 2). The mucosa overlying the swelling appeared normal with no secondary changes. A provisional diagnosis of radicular cyst with 26 was made based on the history and clinical examination findings.

Set of conventional radiographs like intra oral periapical radiograph (IOPAR) with teeth 24, 25, 26, maxillary topographic occlusal, orthopantomogram (OPG) and para nasal sinus (PNS) view were taken.

IOPAR and occlusal radiograph revealed loss of lamina dura and well defined radiolucency extending from 23 to 27 involving the root apices with sclerosing borders. The associated teeth had no significant root resorption (fig 3

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& 4). OPG revealed unilocular radiolucency in an interradicular position between the roots of 23 to 27, lifting of the floor of the left maxillay antrum and displacement of roots of 24, 25 and 27 (fig 5). The PNS view showed uniform haziness in the left maxillary antrum. The left antral floor was intact and lifted superiorly (fig 6). Radiological diagnosis was consistent with the clinical diagnosis of radicular cyst with 26.Fine needle aspiration yielded blood tinged thick, sticky, mucous fluid .The vitality test performed with the teeth 23, 24, 25 and 27 showed that the teeth were non vital.
The patient was subjected for root canal therapy for the teeth 23, 24, 25 and 27 before the enucleation of the cystic lesion.

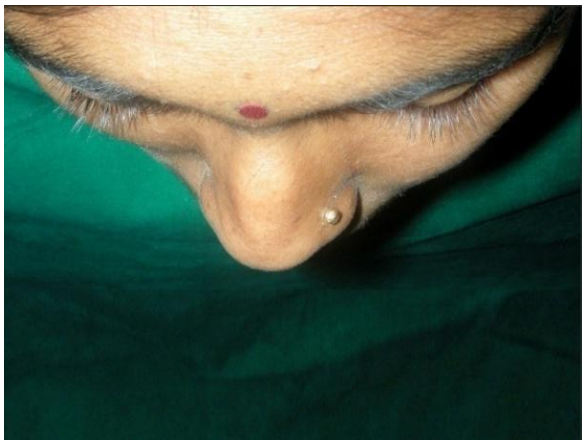

Fig.1 – Diffuse swelling over the left malar region

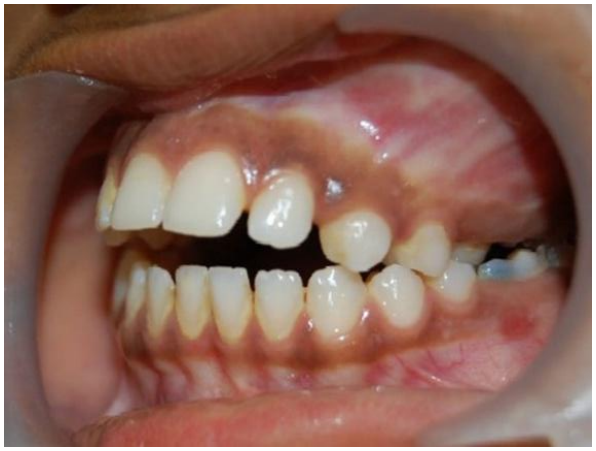

Fig.2 – Vestibular obliteration with respect to teeth 23, 24, 25 extending posteriorly.

The post surgical specimen was studied histopathologically and showed flat interface between epithelium and connective tissue wall. The superficial epithelial cells lining the cyst wall show cuboidal and ciliated epithelium with focal areas of mucous metaplasia. Connective tissue also show chronic inflammatory cell infiltration predominantly lymphocytes and plasma cells (Fig.7).

This was suggestive of glandular odontogenic cyst in left posterior maxilla. The year follow up of the patient showed no recurrence of the lesion.

DISCUSSION

GOC is a rare cyst of the jaw that appears to be a distinct entity because of its unusual histopathologic features. It is suggested that the GOC could be of inflammatory origin, in the presence of chronic apical periodontitis before the cyst[5].

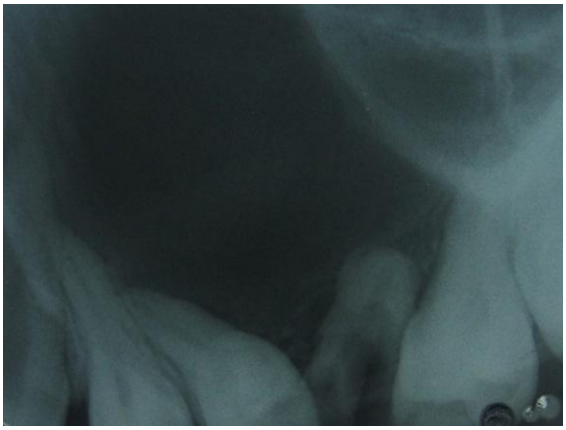

Fig.3 – IOPA revealing periapical radiolucency with 24 extending upto 27 with flared roots of 24, 25 and 27 with a grossly decayed 26.

Some researchers believe that GOC is often misdiagnosed because of the overlap of its histological features with other odontogenic cysts, such as botryoid or lateral periodontal cysts or central low-grade mucoepidermoid carcinoma. Manor et al have reported 56 cases of GOC with the age range of 14 to 90 years (mean age of 50 years), mandible being affected four times than maxilla and predilection for occurrence in anterior regions of the jaw [3]. The lesion in the present case was seen in maxillary posterior region which is relatively a rare.

The female: male ratio is 28:19 [1], showing slightly more predilection for females which was evident in the present case as well. Occasionally it has been described in teenagers [2] as seen in our case.

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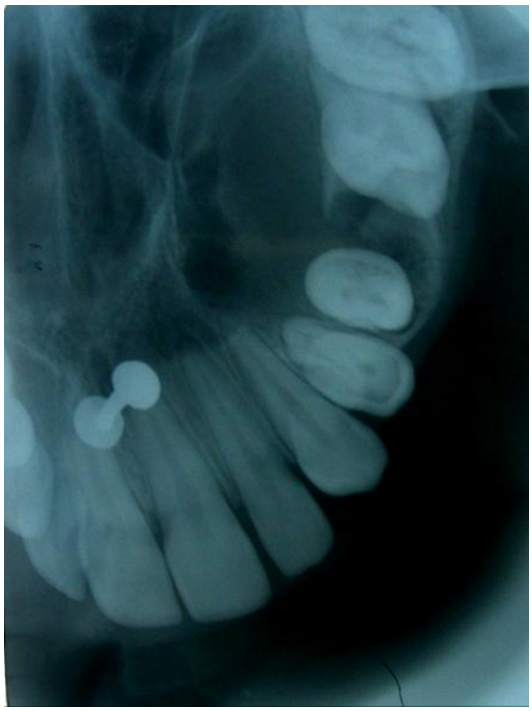

Fig.4 – Occlusal radiograph showing diffuse solitary radiolucency extending from 23-27

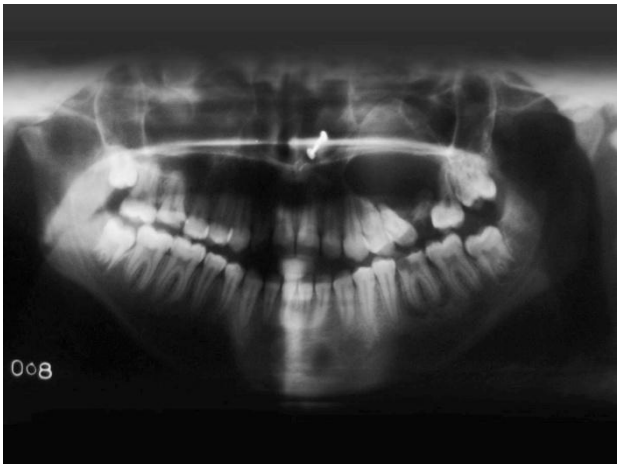

Fig.5 - Unilocular radiolucency in an interradicular position between the roots of 23 -27, with lifting of the floor of the left maxillay antrum and displacement of roots of 24, 25 and 27.

One of the most common manifestations of GOC is a painless swelling of the involved site though, some cases may present with painful swelling due to stretching of or pressure on neurovascular bundles[1]. The radiographic appearance of GOC varies and is not pathognomonic. It may manifest as either unilocular or multilocular radiolucency, usually with well defined scalloped margins often crossing the midline. The locularity (unilocular or multilocular), radiodensity and border characteristics of GOC are important in the differential diagnosis. It usually occurs apical to the teeth showing interdental extensions. The present case had radiological findings of unilocular radiolucency with an interradicular extension between the roots of left maxillary canine and first molar.

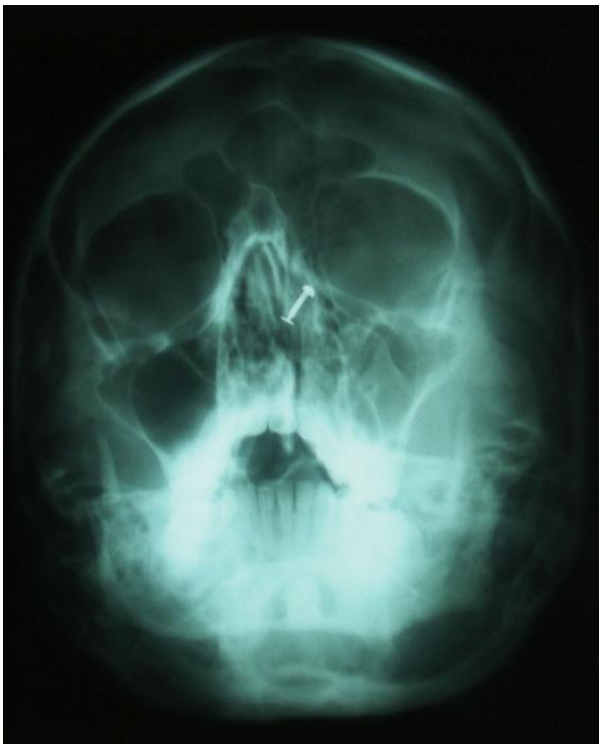

Fig.6 - Uniform haziness, intact floor of the maxillary antrum with lifting of the floor of the left maxillary antrum.

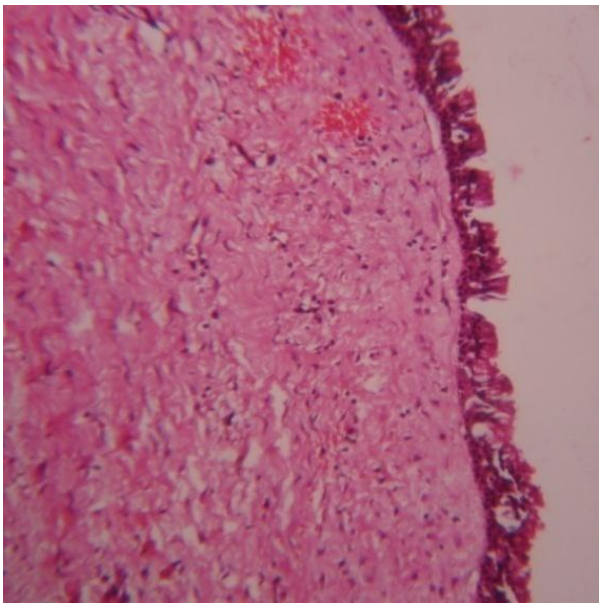

Fig.7 – Flat interface between epithelium and connective tissue wall. The superficial epithelial cells lining the cyst wall show cuboidal and ciliated epithelium with focal areas of mucous metaplasia. Connective tissue also show chronic inflammatory cell infiltration predominantly lymphocytes and plasma cells.

The differential diagnosis of GOC should include few slow growing lesions of the jaws. The pre operative aspiration and fluid inspection is advisable. The presence of water clear, low viscosity fluid content may be a clinical indication of presence of a GOC. However the presence of cholesterol crystals and micro organisms questions the validity of clinical diagnosis of GOC [1]. Histologically, GOC shows some characteristic features such as multicystic process that may be partially lined by non keratinised stratified epithelium. The epithelial lining also occasionally contains eosinophillic cuboidal type cells that may or may not be ciliated [2]. The botryoid odontogenic cyst (BOC) demonstrates similar histopathological features as that of GOC. This suggests that GOC may be a histologic variant of BOC [2].

The diagnosis of GOC can be difficult for two main reasons. Firstly, because of the rarity of the lesion and secondly the oral pathologists have only limited past experience with GOC.

GOC is a relatively rare entity presenting with overlapping clinical and radiographic features. The potentially aggressive and an unpredictable nature of GOC are suggested by its osseous extensions, penetration of cortical bones, locally invasive growth and high recurrence rates following conservative treatment. The present case suggests that GOC has to be included in the differential diagnosis of asymptomatic lesions presenting with unilocular radiolucency should include GOC. Early diagnosis and appropriate therapy for GOC is of paramount importance.

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